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Endometrial Osseous Metaplasia: Presentation of Two Rare Cases Related with Infertility, and Their Hysteroscopic Approaching

CASE REPORT

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Abstract

Objective: To present two cases of patients diagnosed of endometrial osseous metaplasia related with infertility and their hysteroscopic approaching.

Design: Case report.

Setting: Two clinical cases diagnosed and treated at Institutional University Hospital.

Patients: Two premenopausal women diagnosed and treated in our Center.

Interventions: Transvaginal ultrasound scan, diagnostic hysteroscopy and hysteroscopic removal of bone fragments.

Main Outcome Measures: Ultrasound scan during further clinical follow up.

Results: Both patients underwent a diagnostic hysteroscopy, and removal of all the bone fragments with total resolution of their pathology.

Conclusions: Endometrial osseous metaplasia is a rare pathology that should be suspected in patients with a history of secondary in-

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fertility, several miscarriages and curettages, that present with an ultrasound image compatible with an intrauterine device. Hysteroscopic approach is the best diagnostic and therapeutic option.

Keywords
Endometrial Osseous Metaplasia; Bony Intrauterine Tissue; Miscarriage; Infertility; Hysteroscopy; Unadverted Intrauterine Device.

Introduction

Endometrial osseous metaplasia is a rare endometrial disorder that can be related with infertility and recurrent miscarriages. It is usually diagnosed by ultrasound exams, and the gold standard treatment is hysteroscopy.

The first description was made on 1901 by Mayer [1], a German pathologist. Later, on 1923, Thaler [2] related the presence of bony intrauterine tissue with miscarriage history, and de Brux [3] described the first case of osteogenesis outside genital organs on 1956.

Material and Methods

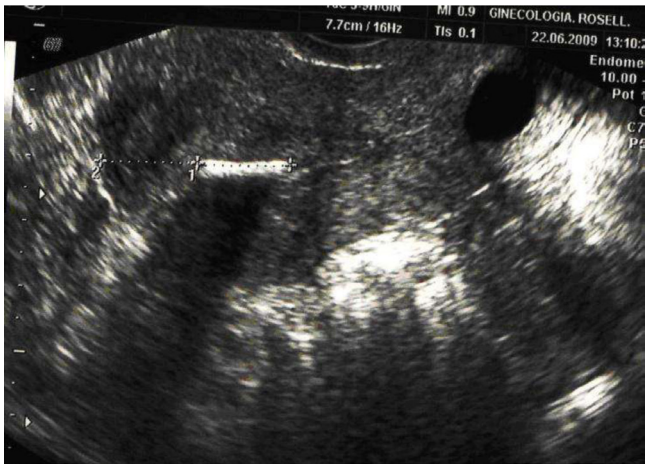
Two cases of endometrial osseous metaplasia diagnosed and treated in our hospital in 2009 and 2010 are presented. Both patients had an infertility clinical history with several miscarriages, suspecting by ultrasound exam the presence of an unadverted intrauterine device. Both patients underwent a diagnostic hysteroscopia, removing all the bony fragments, with complete resolution of the pathology.

Case 1

A 34 year-old woman, was referred to our center because of an unknown intrauterine device finding during an ultrasound exam. The patient had a clinical history of eumenorrhea, without dimenorrhea or dispareunia. The IUD that was found could not be removed at office due to the lack of its tail. The patient had a clinical history of infertility with a miscarriage at the age of 17 and another one six months before this exam. Both miscarriages were treated with endometrial curettage. The ultrasound exam showed a high-refringence intrauterine structure that could correspond with an intrauterine device, slightly shorter than usual **(Figure 1)**

The patient underwent a diagnostic hysteroscopy (Karl Storz 5mm hysteroscope) that showed an apparently normal endometrial cavity, without intrauterine devices. On the posterior wall of the uterus, a white and hard surface was found, and several

Figure 1: High-refringence intrauterine structure that could correspond to an intrauterine device during an ultrasound study.



spiculated, white coloured pieces were removed, similar to fetal bone.

Hystologic study showed the presence of bony tissue surrounded by endometrial stroma and glands, according to the diagnosis of endometrial estromal ossification.

One month after hysteroscopy, ultrasound scan was normal, without evidence of residual calcifications.

Case 2

A 39 year-old woman, referred from our fertility office to perform a diagnostic hysteroscopy. During ultrasound scan of a structure corresponding to an intrauterine device was found, unknown to the patient. This woman had an infertility history of six years, with a previous miscarriage at the age of 29 treated by endometrial curettage. The patient had a history of eumenorrhea, without dysmenorrhoea or dispareunia. Ultrasound exam showed the presence of a 12.9 mm intrauterine device placed 5 mm away from the fundus of the uterus **(Figure 2)**. During hysteroscopy, a coral-like plaque was found on the anterior endometrial surface that could be removed using forceps **(Figure 3)**.

Pathologic study later showed endometrial secreting type structures with stromal ossification areas, finding laminated spongy bone and adipose bone marrow without any evidence of haematopoiesis. Surrounding stroma had no foreign-body reaction or giant cell response, therefore, it was diagnosed as stromal endometrial ossification. **(Figure 4)**.

Figure 2: Scan image resembling unadverted intrauterine device.

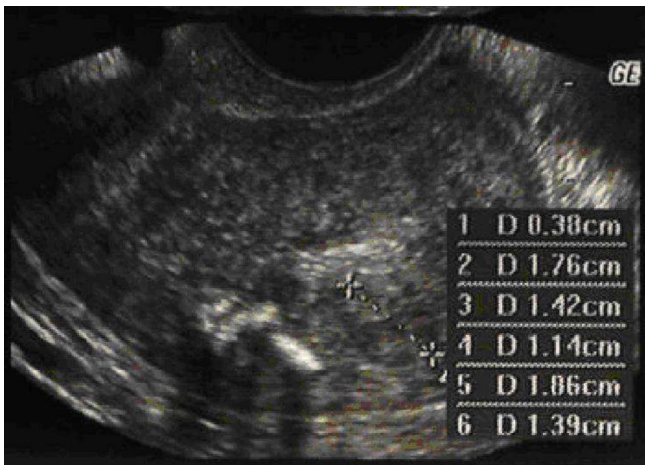


Figure 3: Coral-like plaque image in hysteroscopy.

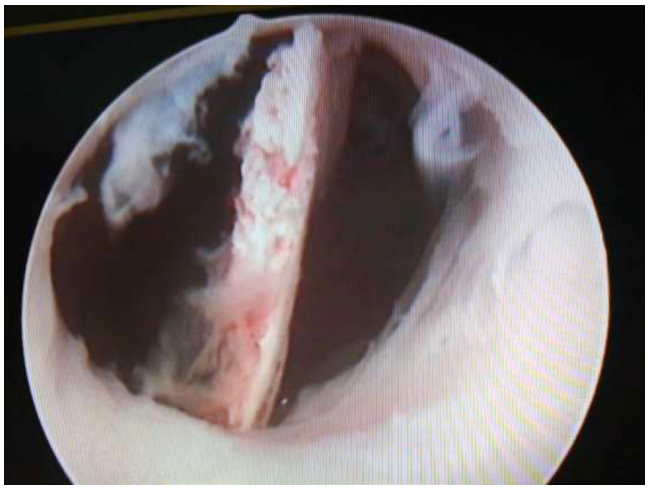
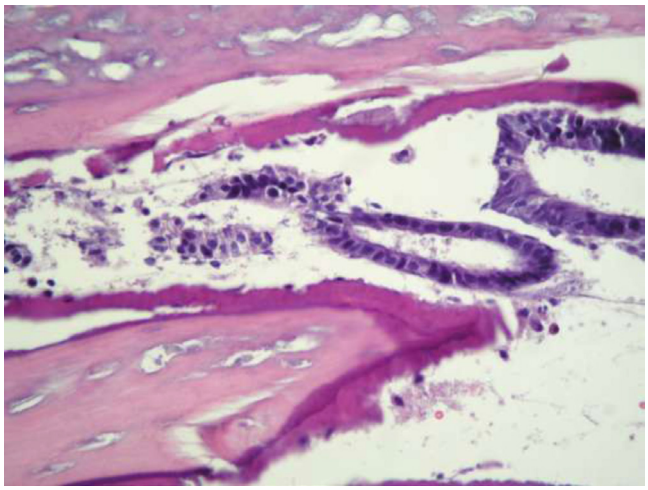


Figure 4: Histological laminated spongy bone and adipose bone marrow without evidence of haematopoiesis. It's possible to observe tubular glands coated with one layered ciliated cubic epithelium without nuclear atypia between trabeculae. That covering epithelium resembles endometrial appearence.



Results

Both patients underwent hysteroscopic treatment to remove all the bony fragments, with complete resolution of their pathology.

Discussion

Endometrial stromal metaplasia includes the formation of nests of smooth muscle, cartilage and bone in the endometrial stroma [4]. The newest World Health Association (WHO) classification divides them into cases with epithelial and cases with non-epithelial changes. Among non-epithelial metaplasia and related changes we can find smooth muscle metaplasia, osseous metaplasia, cartilaginous metaplasia, fatty change, glial tissue or foam cell change.

There are two main pathogenic mechanisms proposed to explain the development of this disorder. There are some authors who propose it is secondary to the retention of fetal bone fragments surrounded by endometrial stroma while other cases are a real stromal metaplasia after a performed curettage [8]. Other authors defend it is a real metaplastic transformation of endometrial stroma with an ex-novo bony creation [9] as a response to an inflammatory reaction secondary to curettage [10] or as a response to chronic endometritis related with it [11].

However, new studies show the existence of osseous metaplasia on several tissues outside of the endometrium, such as breast, cervix or lungs [12, 13, 14]. This finding could be explained by pluripotential capability of stromal cells to transform themselves into bony tissue.

In the case of endometrial stromal metaplasia, this could be a response to chronic inflammation due to a miscarriage or curettage, probably mediated by cytokines. In the same way, chronic endometritis can also stimulate the proliferation of mesenchymal cells that have an inherent capability of metaplasia and can differentiate into chondroblasts or osteoblasts.

We would like to highlight that, regardless of how it develops, osseous metaplastic tissue can be found in relation to malignant Mullerian mixed heterologous tumours, sarcomas and endometrial carcinomas [15].

Presence of bony tissue inside uterine cavity can interfere with embryo implantation and cause infertility due to a significant rising of E2 prostaglandin concentration inside the uterus, as it was demonstrated by Lewis et al [16]. Other authors propose that a reactive endometritis secondary to bony fragments presence could be the cause of infertility, preventing blastocyst implantation [17].

Although histology is the gold standard to diagnose endometrial osseous metaplasia, transvaginal ultrasound examination can show suspicious images sugesting bone presence inside the uterine cavity. These images have low specificity, but sometimes, as both of the cases we describe, can remind us of the presence of an IUD [18], and the patient not knowing of its existence can be the main clue to suspect this entity. Other pathologies that have to be taken into account in the differential diagnosis are Asherman's syndrome, a calcified myoma, intrauterine foreign bodies and heterotopic calcificied materials.

Hysteroscopy is currently the gold standard approach to diagnosis and treatment of osseous metaplasia [5, 9, 17-21], because we can see and remove directly all the bony tissue. In our two cases we used a diagnostic hysteroscope, in one surgical intervention alone. It was enough to see the complete uterine cavity and remove all the bony fragments, as was demonstrated during ultrasound examination afterwards performed. We believe that a surgical hysteroscope should be used, including a resector and endometrial curettage in difficult cases with persistent ossification or symptoms, because there are less adverse effects associated that could affect the patient's reproductive future with hysteroscopic approach than with other types of management .

Conclusion

Endometrial osseous metaplasia is a rare pathology that should be suspected in patients with a history of secondary infertility, several miscarriages and curettages, that present with an ultrasound image compatible with an intrauterine device. Hysteroscopic approach is the best diagnostic and therapeutic option.

Acknowledgement

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